

cases; critical review of the literature; critical review of the manuscript; approval of the final version of the manuscript.

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Conflicts of interest

None declared.

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Lupus miliaris disseminatus faciei with extra-facial involvement in a 6-year-old Japanese girl*



Dear Editor,

Lupus miliaris disseminatus faciei (LMDF) predominantly occurs at 20–30 years of age and is rarely seen in children. We report a pediatric case of LMDF affecting the face and the labia majora.

A 6-year-old Japanese girl was referred to our department, complaining of a 5-month history of pruritic papular eruptions on the face. She had been treated with oral antiallergic drugs and topical corticosteroids, but without effects. Physical examination showed numerous 1–2 mm dome-shaped small reddish papulonodules around the mouth and lower eyelids (Fig. 1 A and B). In addition, reddish papules were scattered in the labia majora (Fig. 2). A skin biopsy was carried out from papular eruptions on the right jaw. Histological examination revealed dermal epithelioid cell granulomas without caseous necrosis (Fig. 3A).

Magnified epithelioid cell granuloma images showed multinucleated giant cells in the dermis (Fig. 3B), as well as inflammatory lymphocytic and histiocytic infiltration around the granulomas and hair follicles area. Immunostaining was positive for CD68 and CD163 antigens (Fig. 3 C and D). A tuberculin test was negative. Treatment with oral administration of clarithromycin showed favorable effects on the vulvar lesions after 5 months and the facial lesions after 9 months.

The present case developed multiple palpebral and perioral papulonodular lesions, which are the frequently involved sites of LMDF. In addition, vulvar involvement was observed. The patient did not have any organ symptoms suggestive of juvenile-onset sarcoidosis. Granulomatous rosacea was excluded, because neither facial erythema nor telangiectasia was observed, and the patient denied flushing.

Clinical and pathological features of pediatric LMDF are slightly different from adult LMDF, such as i) Papules concentrated around the mouth, on the nasolabial fold, and on the lower eyelids, ii) Small papule size, iii) Few pustules and scarring, iv) Redness around the mouth, v) Few caseous necrosis within epithelioid granulomas, and vi) Short clinical course.¹ On the other hand, Childhood Granulomatous Periorificial Dermatitis (CGPD) was reported as a disease in which yellow-brown papular eruptions lim-

* Study conducted at the Fukushima Medical University, Fukushima, Japan.

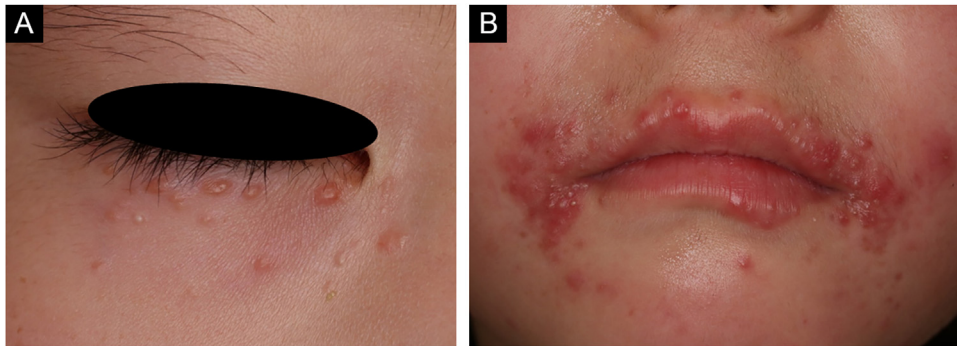


Figure 1 Numerous small erythematous papules were located on the lower eyelid (A) and around the mouth (B).



Figure 2 Reddish papules were scattered in the labia majora.

Table 1 Cases of vulvar involvement in childhood Lupus miliaris disseminatus faciei and childhood granulomatous periorificial dermatitis.

Author and year of publication	Disease	Age in years	Gender	Clinical features	Treatment	Treatment duration
Wataeda et al. (1990)	LMDF in children	9	Female	Face, labia majora	Oral minocycline, topical non-steroidal anti-inflammatory drug	3 mo and a half
Andry et al. (1995)	CGPD	Unknown	Female	Face, perivulvar	Unknown	Unknown
Amy et al. (2002)	CGPD	6	Female	Face, labia majora	Oral and topical erythromycin	2 mo
Amy et al. (2002)	CGPD	8	Female	Face, arms, abdomen, labia majora	Oral erythromycin, topical metronidazole	Unknown

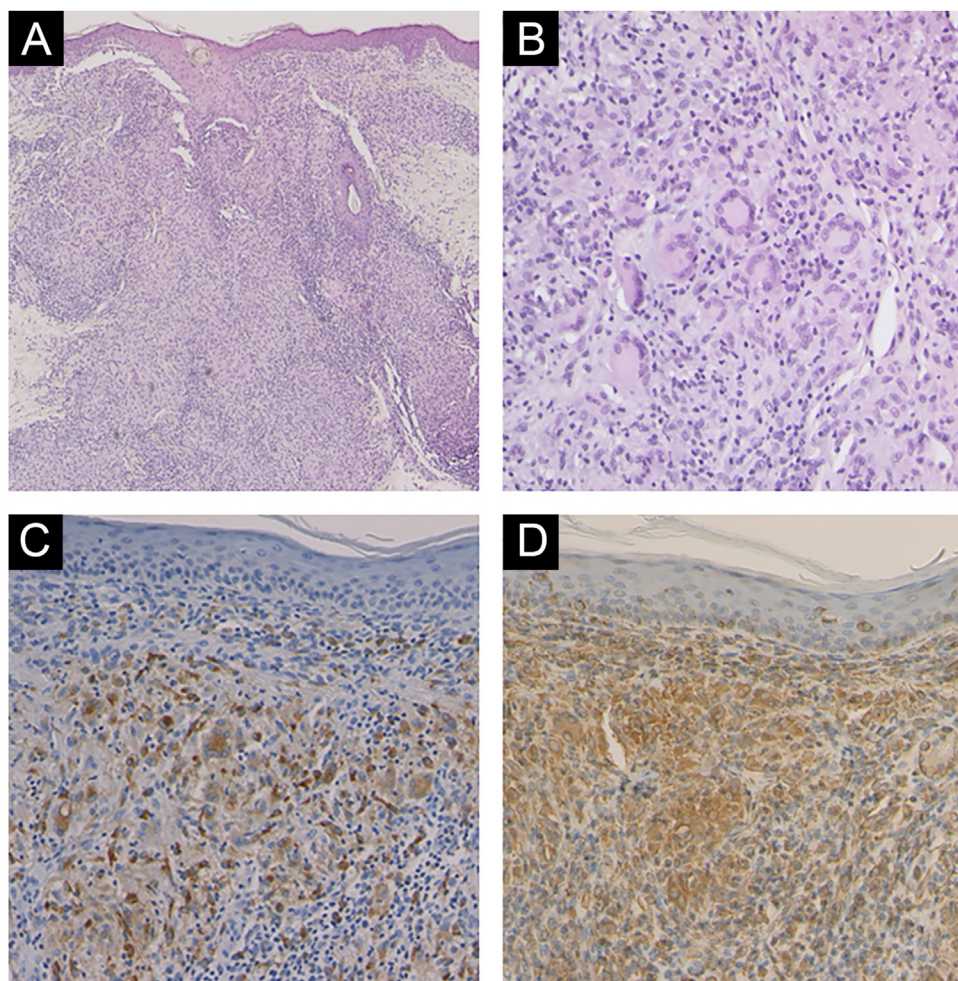


Figure 3 Histopathological features showing epithelioid cell granulomas within the dermis (A). Higher magnification reveals epithelioid cell granulomas containing multinucleated giant cells (B). Immunohistochemistry revealed positive findings for CD68 (C) and CD163 (D).

ited to the perioral, perinasal, and periocular regions that histopathologically show epithelioid cell granulomas around hair follicles.² The features of LMDF in childhood are highly similar to those of CGPD, and differentiation of both disorders is challenging and possibly both disorders are the same entity.¹

There are several cases of LMDF with extra-facial involvement in sites such as the neck, axilla, groin, and extremities. Genital regions are also affected, and to our knowledge, there are only four reported cases of this involvement in childhood LMDF (1 case) and CGPD (3 cases) (Table 1).^{3,4} The age of onset was 6–9 years, and all cases were girls. They were successfully treated with oral minocycline, topical non-steroidal anti-inflammatory drugs, oral and topical erythromycin, topical metronidazole, topical tacrolimus, and topical metronidazole.^{4,5} Treatment response is better in pediatric LMDF than in adult cases. In pediatric cases of LMDF, genital areas should be examined in detail.

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Authors' contributions

Misaki Kusano: Approval of the final version of the manuscript; critical literature review; data collection; analysis and interpretation; study conception and planning; management of studied cases; manuscript critical review; preparation and writing of the manuscript.

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Conflicts of interest

None declared.

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Paraneoplastic acquired ichthyosis as the first manifestation in breast implant-associated anaplastic large cell lymphoma*

Dear Editor,

Anaplastic large cell lymphoma (ALCL) accounts for 1%–3% of non-Hodgkin's lymphomas and approximately 15% of T-lymphomas. The 5th edition of the World Health Organization (WHO) classification of hematolymphoid tumors recognizes four different subtypes of ALCL: ALK-positive, ALK-negative, primary cutaneous, and those associated with breast implants (BIA-ALCL).

In January 2011, the Food and Drug Administration (FDA) published 34 cases collected between 1997 and 2010, with a probable association between ALCL and the use of breast implants. Owing to the implementation of the PROFILE registry, approximately 900 cases have been reported worldwide.

Implants are categorized according to their internal fill (saline or silicone), shape (symmetric or asymmetric), or external surface (smooth or textured).^{1,2} Textured implants have the greatest association with this type of lymphoma.

We present the case of a 43-year-old woman with a history of bilateral breast prosthesis implantation (silicone-textured), 6-years ago for aesthetic reasons. She visited the clinic because of the presence of scaly plaques with cracks and erythematous edges throughout her body, with a 3-month history. In some plaques, fine superficial scaling was prominent in the intermammary fold, scalp, and on the edge of the eyelids. Physical examination revealed a fixed, painless, 1.5 cm adenopathy in the right axillary region (Fig. 1).

Chest CT scan, along with a whole-body PET/CT, revealed a liquid collection surrounding the right mammary implant, with an increase in soft tissues in the chondrosternal joint, poor definition of the pectoral planes in its medial portion,



and trabeculation of subcutaneous fat. In contrast, multiple pathological adenopathies were observed in the anterior mediastinum, bilateral axillary regions, and supraclavicular fossae (Fig. 2A–C).

While a skin biopsy was suggestive of ichthyosis, subsequent biopsy of the right axillary lymph node revealed neoplastic cells with a multilobulated anaplastic morphology and an “embryoid” appearance with foci of tumor necrosis. Immunohistochemistry phenotypes were CD45+ CD30+, CD43+, Bcl-2+, MUM-1+, MIB-1+(60%), CD3-, CD20-, CD79a-, CD10-, BCL6-, CD38-, LMP1-, ALK-, suggestive of anaplastic large cell lymphoma (Fig. 3A–F).

Based on the clinical, radiological, and pathological findings, the diagnosis of BIA-ALCL with a locally aggressive or extensive stage was established (stage III, T4N2M0).

BIA-ALCL commonly presents as a late peri-implant seroma (>80%), causing distortion and asymmetry of the breasts. It can manifest with regional lymphadenopathy (predominantly axillary, supra-, or infra-clavicular) in 20% of cases, or concurrent with skin lesions (erythema, rash, erythematous nodules) or as de novo in 8% of cases.^{3–7}

In our case, ichthyosis was the initial cutaneous paraneoplastic manifestation of BIA-ALCL and dermatological management was based on glycerin soap, liquid petroleum jelly, and methylprednisolone aceponate (ointment). This cutaneous alteration can appear in the same manner in endocrinopathies, immunological disorders, vitamin deficiencies, infections, and with drug use.³

BIA-ALCL originates from the fibrous capsule surrounding the implant, except in the advanced stages, which involve the surrounding tissue. Stage I was diagnosed in 83% of patients, and only 7% presented with stage IV disease.^{1,6}

The etiopathogenesis of BIA-ALCL is multifactorial, namely chronic infection in the textured implant configuration, gram-negative biofilm formation, immune response, chronic inflammation, host genetics (e.g., JAK/STAT, p53), and timing of oncogenesis. The presumptive triggers include mechanical friction, silicone implant shell particles, silicone leachate, and bacteria. BIA-ALCL presents at a median time of 8–10 years after breast prosthetic implantation.^{6–9}

Considering that 2%–4% of cases can occur bilaterally, both implants of our patient were removed with

* Study conducted at the 12 de Octubre Hospital, Madrid, España.